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## Cavernous Lymphangioma of the Breast: Case Report of an Infant

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### Summary

Ultrasonography and magnetic resonance imaging (MRI) were used to assess a 4-month-old male infant with a multicystic tumor of the left breast that was identified at birth. The tumor was removed surgically following the diagnosis of a cavernous hemangioma. Only seven similar cases of lymphangioma of the breast have been reported. We review these cases in conjunction with our own findings.

### Introduction

Lymphangiomas are thought to be benign lymphatic tumors, which usually arise in the necks and axillae of children. Lymphangiomas arising in the breast region are rare and only seven cases have been reported<sup>1-7)</sup>. Lymphangiomas in adults are even less common<sup>6)</sup>. Ultrasonography, magnetic resonance imaging (MRI), cystography, and computed tomography (CT) can facilitate the diagnostic process and are useful for preoperative assessment. We present a case of an infant with a cavernous lymphangioma of the left breast who, following ultrasonography and MRI examinations, had the tumor surgically excised.

### Case report

A 4-month-old male infant was admitted to the National Sendai Hospital with a soft tumor in his left breast which had been identified at birth. Several needle biopsies of the tumor had been performed at a rural hospital and clear straw-colored fluid was aspirated on each occasion. However, as the tumor gradually enlarged, sometimes the fluid appeared sanguineous, so he was transferred to our hospital for further treatment.

A soft, round, hen-egg size tumor was located in the patient's left breast, immediately below the areola mammae (Fig. 1). Ultrasonography displayed a multicystic tumor, with an anterior cyst located in close contact with the skin of the areola. However, the parenchymal part of the tumor borders were unclear. Magnetic resonance imaging also revealed a multicystic tumor in the left

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Key words: Cavernous Lymphangioma, Breast tumor.

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breast without extension into the pleural cavity (Fig. 2).

A diagnosis of cavernous lymphangioma of the breast was made from the history and from these imaging studies, and a 7 × 6.5 cm tumor was excised on August 10, 1993. The tumor had invaded the pectoralis major muscle. Approximately 3 ml of straw-colored fluid was aspirated from the largest cyst. Microscopic examination of the tumor disclosed that the appearance of the tumor was

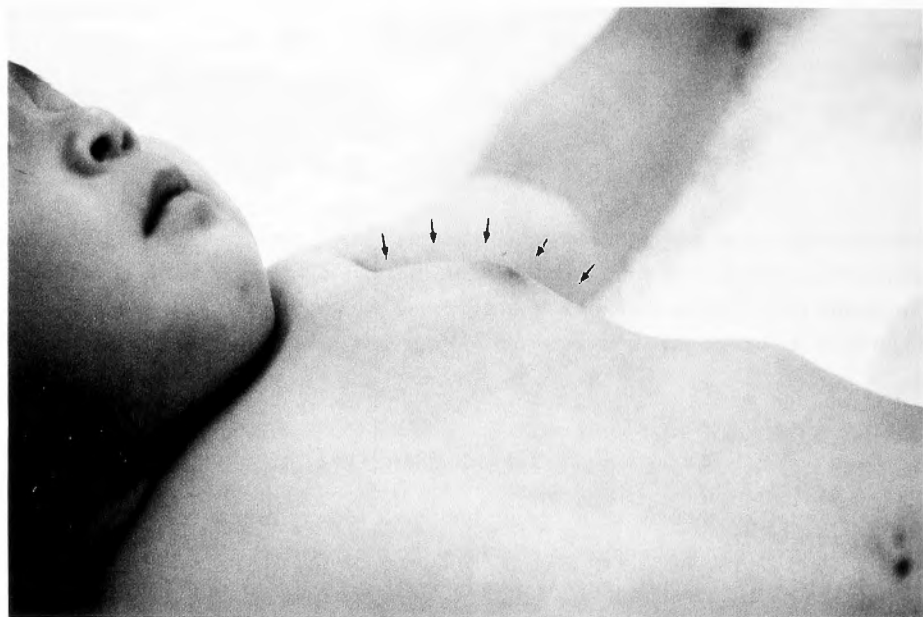


Fig. 1 Photograph of the patient demonstrating the protruded left breast (arrow).

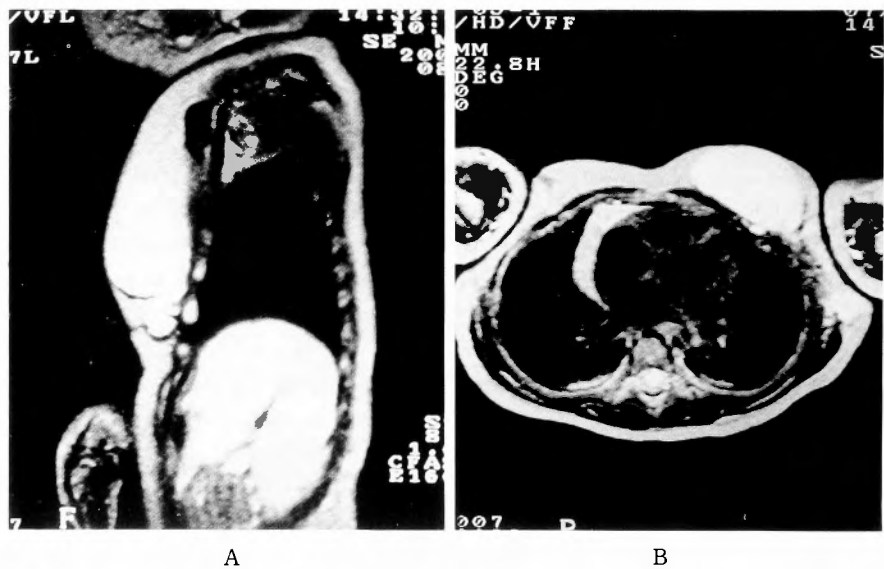
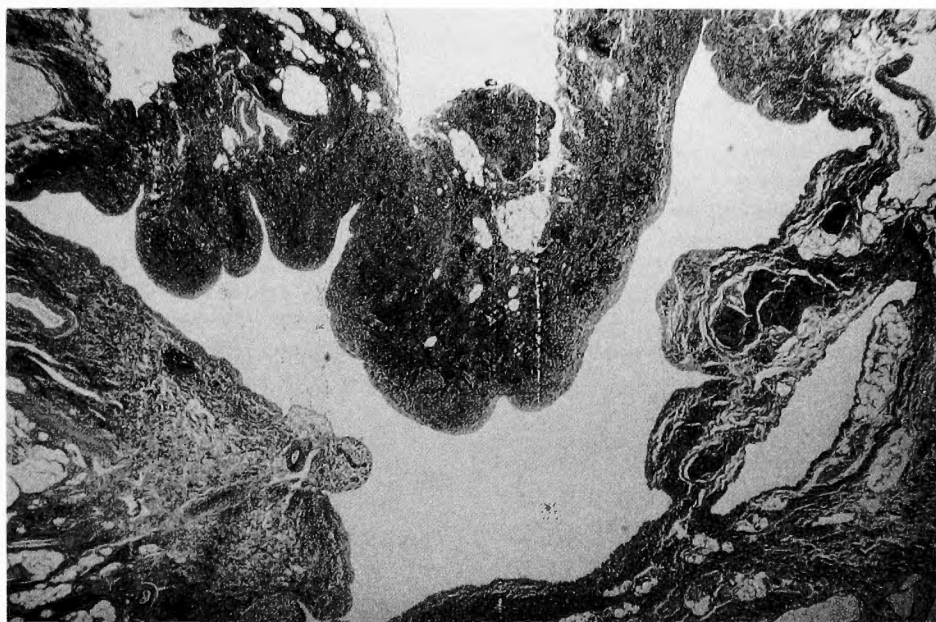


Fig. 2 MRI demonstrating multicystic tumor of the breast. A: sagittal section, B: transverse section.



**Fig. 3** Photomicrograph of the tumor showing dilated lymphatic channels with fibrous septa.

consistent with that of a cavernous lymphangioma (Fig. 3). The patient's postoperative course was uneventful.

### Comment

Lymphangiomas are composed of cystic spaces lined with endothelial cells and frequently arise in the necks and axillae of children. Few cases of the lymphangiomas arising in the breast region have been reported in children or adults<sup>1-7</sup>). Although cystic lesions of the breast are observed frequently in adult patients with fibrocystic disease, they are rarely demonstrated in children. The cavernous lymphangioma consists of dilated lymphatic channels with fibrous septa and penetrates contiguous muscle tissue. This tendency makes it all the more difficult to excise it without sacrificing normal tissue.

There appears to be no significant differences in the side of the involved breast, although only the left breast has been affected in children<sup>2,7</sup>). Reported adult cases have been limited to woman<sup>1,4,5</sup>), while child cases only have involved males.

The diagnosis of lymphangioma of the breast is rarely difficult in children, although the incidence is low. Ultrasonography, CT, and MRI are useful both for diagnostic purposes and for preoperative assessment. In the breast region, punctate analysis of the cystic tumor, or cystography, facilitate diagnosis<sup>4</sup>). Generally, cavernous lymphangiomas do not form definite tumors but consist of diffuse proliferations<sup>6</sup>). Therefore, care should be taken to preserve as much of the normal mammary tissue as possible, especially in cases involving women. We recommend that only definite cystic part should be removed surgically in children even if the tumor is incompletely excised. Sclerosing therapy combined with surgery, or multiple operations over several years are occasionally necessary to achieve an optimal result. There have been no reports of malignant cavernous lym-

phangiomas.

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和文抄録

## 乳房部海綿状リンパ管腫の1乳児例

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生下時よりみられた左側乳房部のリンパ管腫を治療する機会を得た。来院時は4カ月(男児)で既に数回の穿刺が行なわれていたが、腫瘍が増大し、実質性の部分があり、また内容が血性となってきたため手術を目的として当科に紹介された。MRIを中心とした検査を行ない、手術的に摘出し、組織検査で海綿状リン

パ管腫であることを確認した。乳房部のリンパ管腫はこれまで7例が報告されているが小児の例はいずれも左側で、生下時よりみられ、組織学的には海綿状であり、OK-432の局所注入よりも摘出が望ましいと思われるが、出来るだけ乳腺組織の温存をはかるべきである。